

CASE REPORT

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Effort thrombosis: Paget–Schroetter syndrome in a 19-year-old female

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ABSTRACT

Paget–Schroetter syndrome (PSS) is a form of effort thrombosis seen in young athletes and individuals with occupations involving repetitive upper limb movement. Arm swelling, distended veins, skin discoloration, and pain are characteristics of it. Diagnosis can be made with non-invasive investigations, such as Doppler ultrasound; however, a computed tomography venogram, magnetic resonance venogram, or traditional venogram may be required. Treatment involves anticoagulation, thrombolysis, and surgical decompression with removal of the first rib, and this is usually met with good outcomes. We present a young female diagnosed with PSS and requiring a staged multidisciplinary approach to treatment.

Keywords: Effort thrombosis, Paget–Schroetter syndrome, Venous thrombosis

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INTRODUCTION

Paget–Schroetter syndrome (PSS) was named after James Paget and Von Schroetter, who first described and identified its etiology in the 1800s. Paget–Schroetter syndrome, also known as effort thrombosis, is a form of spontaneous thrombosis involving the axillary and subclavian veins, usually following a repetitive force on the vessel as it passes through the narrow space between the clavicle and the first rib in the anterior part of the thoracic outlet [1]. It has an incidence of 1–2 individuals/100,000/year and occurs more commonly in individuals in their 20s–30s. It is also more typical in males, with a male:female ratio 2:1 [2]. It is a venous variant of thoracic outlet syndrome [3, 4]. Other causes of upper extremity deep venous thrombosis include thrombophilia, the use of central venous catheters, trauma, and malignancy.

CASE REPORT

A 19-year-old right-handed female with a history of migraine presented to the Emergency Room with the acute onset of pain, swelling, and bluish discoloration of her right arm (Figure 1). Swelling and discoloration were present throughout the day. There was associated numbness and prominent distended veins in her right arm. There was no antecedent trauma and no personal or family history of venous thromboembolism. The patient works as an aesthetician and engages in repetitive activities such as waxing; she was also a cheerleader for ten years.

Her vital signs were heart rate (HR) 97 bpm, blood pressure (BP) 130/70 mmHg, respiratory rate 18 cpm, and temperature 36.1 °C. She had a positive Urschel's sign (differential swelling with prominently distended veins and red discoloration of the skin) over the right upper extremity, but peripheral pulses were intact. Adson test, supraclavicular compression test, Wright test, and costoclavicular maneuvers were negative. Her labs were unremarkable.



Figure 1: Picture of patient's upper extremities, red arrow pointing to right forearm showing differential swelling, discoloration, and distended veins.

Right upper extremity duplex ultrasound in the Emergency Room did not reveal deep vein thrombosis. Despite negative special tests for thoracic outlet syndrome (TOS), the patient's symptoms were suspected to be due to TOS. She was seen by Vascular Surgery four weeks after the onset of her symptoms, and her repeat duplex ultrasound showed thrombosis in the right subclavian vein. A right upper extremity venogram showed thrombosis of the brachial vein in the upper arm near the confluence with the basilic vein, with multiple venous collaterals in the right arm at the shoulder level. There was also an appearance of chronic thrombosis of the right subclavian vein beneath the clavicle with collaterals, but the superior vena cava was patent. A thrombus was seen in the axillary vein at the level of the proximal humerus and in the subclavian vein beneath the clavicle on intravascular ultrasound (IVUS). She was admitted and underwent catheter-directed thrombolysis with tissue plasminogen activator (tPA), with improvement in the upper extremity collaterals but no significant improvement in her subclavian vein stenosis.

A repeat right upper extremity venogram showed a patent previously thrombosed basilic and axillary vein. The right subclavian vein's patency was restored following suction thrombectomy and balloon angioplasty. The patient had a right first rib excision in that same admission and was discharged home on a three-month course of anticoagulation with rivaroxaban.

DISCUSSION

Our patient presented with acute right upper extremity swelling, discoloration, and distended veins that were suspicious for thoracic outlet syndrome, but her examination findings were inconsistent with TOS. She had been a cheerleader for almost 10 years, and her current job as an aesthetician entailed several hours of waxing daily, an activity that involves repetitive hyperabduction of the shoulder.

Pathophysiology

A causal relationship has been reported between sporting activities leading to hyperabduction, retroversion, and PSS. These movements cause repetitive trauma to the venous intima as it passes through the tight thoracic outlet, leading to activation of the coagulation system and consequent thrombosis [4]. An abnormal congenital lateral insertion of the costoclavicular ligament on the first rib may cause venous occlusion and thrombosis in PSS [3, 5]. Some studies suggest a significant number of patients with PSS have underlying thrombophilia [6–9], although it is not clear if this is a causal or casual relationship. Cassada et al. found an increased risk of postoperative complications in patients with concurrent PSS and thrombophilia [10]. These claims were later disputed.

Paget–Schroetter syndrome has a predilection for healthy young men, but it can also occur in females. The common denominator in both genders is “effort,” or activities that cause trauma to the subclavian vein.

Clinical features

Presenting symptoms include but are not limited to swelling and heaviness, red or bluish discoloration of the arm, and dilated, visible veins across the shoulder and upper arm (Urschel's sign). The trigger may not be immediately apparent, but some patients will report an onset following a specific sporting activity. Clinical signs and symptoms are suggestive, but alone, they have poor specificity in diagnosing PSS [5].

Diagnosis

Doppler ultrasonography is the first-line diagnostic investigation, with a sensitivity of 56% to 100% and specificity of 77% to 100% in diagnosing upper extremity deep venous thrombosis [11]. The variability in sensitivity and specificity is hypothesized to be due to differences in patient selection, the ultrasonographic technique used, and the anatomical location of the subclavian vein under the clavicle. It is not surprising that our patient had a falsely negative study on her first Doppler ultrasound, which could be attributed to an early non-occlusive thrombus. Contrast venography is the gold standard for diagnosing Paget–Schroetter syndrome. Computed tomographic venography (CTV) and magnetic resonance

venography (MRV) are being increasingly used, especially in patients with high clinical suspicion, because they are less invasive and have similar sensitivity to conventional venography [5]. Since the role of thrombophilia in PSS remains uncertain, selected patients should be screened for coagulation abnormalities, as this may influence the need for long-term anticoagulation [5]. Our patient underwent thrombophilia screening prior to the commencement of anticoagulation. Testing for proteins C and S, factor V Leiden, prothrombin gene mutation, and antiphospholipid antibodies returned negative.

Treatment

There are varying opinions on the ideal treatment. An aggressive treatment strategy that involves thoracic outlet decompression, thrombolysis, thrombectomy, percutaneous and surgical venoplasty, venous bypass, and stent placement has an almost 100% success rate. Catheter-directed thrombolysis was employed in this patient, offering a therapeutic effect similar to systemic thrombolysis without systemic side effects. This was followed by thoracic outlet decompression (TOD) surgery and oral anticoagulation. The higher incidence of post-thrombotic syndrome and recurrent upper extremity deep vein thrombosis (UEDVT) in patients who have delayed TOD surgery favors the current practice of routine and early surgery [12–14]. There is no consensus on the duration of anticoagulation in patients with PSS without underlying predisposing factors, such as thrombophilia, central venous catheters, or lines. Some authors suggest anticoagulation for 3–6 months; others recommend no anticoagulation in patients with good surgical outcomes [3, 14]. Anticoagulation without thrombolysis or first rib resection increases the risk of recurrent symptoms and residual deficits [15].

In a comprehensive study conducted by Spencer et al., 32 competitive athletes with effort thrombosis achieved a favorable outcome with a staged multidisciplinary approach [12]. The first stage is early diagnostic venography and thrombolysis. This is followed by thoracic outlet decompression and, lastly, temporary postoperative anticoagulation.

Like most cases of upper extremity deep venous thrombosis, PSS may be complicated by pulmonary embolism and post-thrombotic syndrome, the latter being more common in PSS [16]. Our patient did well after staged procedures; Physical Therapy cleared her for return to work. At her follow-up visit, she demonstrated significant improvement in swelling and skin discoloration.

CONCLUSION

Paget–Schroetter syndrome is relatively uncommon; it may be associated with a debilitating disability if misdiagnosed. Early diagnosis, prompt thrombus

removal or dissolution, and resection of the 1st rib are crucial to achieving a good outcome following diagnosis. Despite a negative Doppler ultrasound, physicians should identify and proceed to venography, CTV, or MRV in patients with high clinical suspicion of the syndrome. A staged, structured treatment protocol ensures optimal outcomes while minimizing long-term complications in patients.

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Author Contributions

Omobolanle Adetimehin – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Ibiyemi Oke – Conception of the work, Acquisition of data, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the

work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Guarantor of Submission

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Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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